Leucoplakia Buccalis et Lingualis, or Ichthyosis Linguæ; Successful Treatment with the Galvano-Cautery.

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LEUCOPLAKIA BUCCALIS ET LINGUALIS, OR ICHTHYOSIS LINGUÆ;

SUCCESSFUL TREATMENT WITH THE GALVANO-CAUTERY*.

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During the past year it has been my good fortune to cure a case of the rare and intractable disease which forms the subject of this paper.

This disease has been recognized but a short time, and very little can be found upon the subject in general medical literature, but, by a thorough search in the library of the Surgeon-General's Office in Washington, I found about fifty papers, mostly reports of cases, which, with the single exception of one by Bazin, which I take second-hand, seem to embrace all that has been written concerning the disease.

The literature is involved in what at first seemed inextricable confusion, for the reason that several different affections have been confounded and described under the same name. Thus, "smokers' patches," the peculiar condition of the mucous membrane of the mouth found in old glass-

* Read before the American Laryngological Association, June 24, 1885.

blowers, termed by Guinaud * "professional patches," psoriasis linguæ, and various manifestations of syphilis, have been considered by different authors as true leucoplakia, or as one of its phases.

The term leucoplakia, which, of the names proposed for this affection, seems open to least objection, was suggested by Professor Schwimmer,† who gives a lengthy and exhaustive description of the disease. Ullmann ‡ employed the term tylosis, which is strongly advocated by W. Fairlie Clarke.# The affection was first brought prominently before the profession by J. W. Hulke,‖ under the title ichthyosis linguæ, a name which has been quite generally accepted save by Debove ^ and some other French writers, who have described it, together with other affections, as a manifestation of psoriasis.

Definition.—Leucoplakia buccalis is a chronic affection of the buccal mucous membrane, characterized by thickening of the epithelium, and the formation of white, opaline, elevated patches, which usually become fissured and painful, and, after continuing for a long time, are likely to terminate in epithelioma.

History.—The first mention that I can find of ichthyosis linguæ is in a paper under the title of "Maladies de la peau," 1822, by Alibert, who records a case, reported in some of the journals, of a young woman in Naples who had extensive ichthyosis of the skin, which finally involved the tongue and lips. In 1837 Samuel Plumbe ◊ described a

* "Syphilis des reniers," "Lyon méd.," xxxv, 1880.

† "Vierteljahresschrift für Dermatologie," iv, 1877.

‡ "Aerztlich. Intelligenzblatt," Munich, v, 1858.

"British Med. Jour.," 1874, vol. i.

‖ "Med. Times and Gazette," London, 1865, vol. i.

^ Paris thesis, 1873.

◊ "Diseases of the Skin," Philadelphia, 1837.

case of enlargement of the papillæ of the tongue which he regarded as precisely similar in its nature to local ichthyosis of the skin. In 1858 Ullmann * described a case of leucoplakia under the name of tylosis. In the same year J. J. Buzenet † reported two cases of undoubted leucoplakia, and, in 1861, J. W. Hulke ‡ recorded a case under the title of "Wart and Corn of the Mucous Membrane of the Tongue." The following year Sir William Fergusson # presented a somewhat similar case in a clinic at the King's College Hospital, and in the same year, in the report of a clinic by Dr. Andrew Clark, ‖ we find a case described under the name of aphtha figurata, which is supposed by some to have been true leucoplakia, but which seems to have been psoriasis of the tongue. The same year J. Moore Neligan ^ notes a case of true leucoplakia which finally terminated in epithelioma. February 28, 1865, J. W. Hulke, ◊ of London, presented what is generally credited as the first paper on the subject to the Royal Medical and Chirurgical Society. Subsequently brief articles appeared on the subject by S. James A. Salter, ↕ 1866; Bazin, ↕ 1868; M. Lailler, ⇞ 1869; Sir James Paget, ** 1870; C. Saison, †† 1871; and W. Fairlie Clarke, ‡‡ 1872. Finally, in 1873, Charles Mauriac ## published an exhaustive

* *Loc. cit.* † Paris thesis, 1858.

‡ "Med. Times and Gazette," London, 1861, vol. ii.

"Lancet," London, 1862.

‖ "Med. Times and Gazette," London, 1862, vol. ii.

^ "Notes of an Unusual Abnormal Condition of the Mucous Membrane of the Tongue and Cheeks considered in Connection with Life Assurance," 1862.

◊ *Loc. cit.* ↕ "Papillary Tumors of the Gums," 1866.

↕ Paris thesis, 1873.

⇞ "Dict. encycl. des sci. méd.," Paris, 1869.

** "Case of Cancer following Ichthyosis of the Tongue," 1870.

†† Paris thesis, xlvi, 1871. ‡‡ "Lancet," London, 1872, vol. i.

"L'Union médicale," xvi, 1873.

article on the subject under the title of "Du psoriasis de la langue et de la muqueuse buccale." Since Mauriac's article numerous papers have appeared, the most important of which are by M. G. Debove,* 1873; Henry Morris,† 1874; W. Fairlie Clarke,‡ the same year; Robert F. Weir,# 1875; Professor Ernst Schwimmer,‖ 1877 and 1881; and E. Vidal,△ 1883.

The disease is very rare, but its frequency can not be accurately determined, on account of the diversity of opinion which has been entertained by different writers with reference to its ætiology and diagnosis—a diversity which has caused many cases to be placed in this class which do not properly belong in it. Professor Schwimmer saw only twenty examples out of five thousand cases of diseases of the skin and syphilis, and many others have not seen so large a proportion as this; but doubtless many cases are not recognized, and pass for constitutional syphilis.

Most of the cases have been reported from Germany, France, and England, but some have been recorded in this country. So far the records show that the disease seldom affects any excepting males over forty years of age, though it has been observed as early as the thirty-second year, and a very few examples have been seen in women.

Anatomical Characters. — The patches are generally found on the dorsum of the tongue or the inner surface of the cheek and lips, but seldom, if ever, on the lower surface of the tongue or behind the anterior pillars of the fauces, and they are limited to the buccal cavity.

They may be seen in one or more small, irregular, or

* Paris thesis, 1873.

† "British Med. Jour.," 1874, vol. vi.

‡ *Loc. cit.*

"N. Y. Med. Jour.," vol. xxi, 1875.

‖ *Loc. cit.*, and "Transactions of the International Med. Cong.," London, vol. iii, 1881.

△ "L'Union méd.," xxxv, 1883.

oval patches, or these may have become confluent. A considerable portion of the tongue alone may be involved, or the dorsum of the tongue, the buccal mucosa, and the surface of the jaw may one or all be affected.

The first appearance of the white patch is preceded by hyperæmia, and in the early stages a hyperæmic areola is found about its borders. Afterward the patch itself is more or less thickened, sometimes to the extent of six or eight mm., and the epithelium, which has become hard and dry, may be easily removed, or in spots it may have been spontaneously exfoliated, leaving the appearance of an ulcer.

The surface of the patch is marked by numerous fine lines or furrows, which intersect each other, dividing it into small polygonal spaces. Some of these may extend as deep fissures down through the thickened epithelium, and involve the mucosa in painful ulceration. In cases of long standing the papillæ may be much enlarged, giving the surface a warty appearance.

Under the microscope the epithelium is found greatly thickened, the papillæ enlarged and flattened, the blood-vessels dilated, with an accumulation of leucocytes about their walls. The superficial layer of the mucous corium is infiltrated with embryonic cells, and the deep layer is involved in vascular alterations.

According to Hulke,* there is hypertrophy of the epithelial and papillary layer of the mucous membrane, similar to the condition of the skin termed ichthyosis.

In 1874, W. Fairlie Clarke† spoke of the disease as a chronic inflammation and papillary growth, but in later writings he does not strictly adhere to these views.

Mauriac‡ says "it is certain that it is a chronic inflammation of the parts involved."

In the first stage of the affection, hyperæmia of the

* *Loc. cit.* † *Loc. cit.* ‡ *Loc. cit.*

deeper layers of the epithelium exists, which is soon followed by exudation, and this consolidating causes induration.

The epithelial cells are increased and the papillæ enlarged, but later, when the affection merges into epithelioma, there is, according to Clarke,* an enormous increase of the rete mucosum at the expense of the papillæ, which are then reduced to mere threads.

Causation.—Excessive tobacco-smoking is ranked as one of the most frequent causes of the disease, but it is barely possible that prolonged irritation of any character may have a similar effect on those predisposed to it.

Thus, Mauriac † and Vidal ‡ mention highly spiced foods and alcoholic stimulants as irritants which must be avoided.

Schwimmer # calls the affection idiopathic, but both he and Mauriac think there must be some peculiar predisposition toward it in order that it may be developed.

Bazin,‖ who has seen the affection in several members of the same family, believes that it is often, if not usually, the result of constitutional syphilis.

Mauriac believes that all psoriasis of the mouth and tongue supervening in syphilis is not necessarily syphilitic, and may not take part in the syphilitic disease.

Debove, Bazin, and Mauriac attribute it frequently to the arthritic or dartrous diathesis. Thus it will be seen that the cause of the affection is not definitely known, though it is commonly believed to be induced by smoking in most instances. It must not be forgotten, however, that several cases have been recorded in persons not addicted to the use of tobacco. In those who use tobacco to excess it is not necessarily the irritant effects of the smoke or heat that cause the disease, but, as in the case here appended, the noxious influence of the tobacco itself.

* *Loc. cit.* † *Loc. cit.* ‡ *Loc. cit.* # *Loc. cit.* ‖ *Loc. cit.*

Clinical History.—It is also difficult to determine exactly the duration of the disease, for it has generally been discovered accidentally; but usually it will be found to have existed for months or years when the patient first presents himself. This is due to the fact that at first the affection causes no inconvenience. Usually the small patch first observed gradually increases in size until at length stiffness of the part occurs or painful fissures form, which cause the patient to seek advice.

In some cases the epithelial cells gradually pile up until a thick, horny mass is formed, which may then be thrown off spontaneously or pared off by the patient as he would cut off a corn or wart. After a time, varying from a few months to many years, the formation of fissures and ulcers causes pain, and finally, in a large percentage of cases, epithelioma results and runs its usual course. Sometimes the affection will remain stationary for months, or, under the influence of some irritant, it may rapidly progress, but may again become dormant if the irritant is removed. Occasionally unchanging erythematous patches remain in the surrounding mucous membrane for years.

In the cases associated with syphilis and in those that have developed into epithelioma the parts become greatly swollen, and deep, fungous ulcers occur which may erode vessels and cause serious hæmorrhage. In these same cases the lymphatic glands are involved, but this does not occur in the earlier stages of idiopathic leucoplakia.

Often the first symptom noticed by the patient is simply an uneasy sensation; but this may not appear until the disease has existed for years. In other cases the mucous membrane early becomes sensitive, so that spices, hot food or drinks, alcoholics, tobacco, etc., cause more or less pain. When deep fissures occur, the pain may become intense and almost constant, though in some cases it is present only at

intervals. There are no constitutional symptoms until epithelioma is developed. Late in the disease, speaking, mastication, and swallowing sometimes become difficult, especially when epithelioma occurs. In such cases there is also profuse and very troublesome salivation, which continues both night and day.

Upon examining the mouth in the early stages, several more or less oval red or white patches are usually found which are apt to be mistaken for secondary syphilis. These in time become bluish, and finally, with increase in their epithelial covering, grayish or of a milky white color—like mucous membrane touched with nitrate of silver. These spots may remain distinct for a long time, but with the progress of the disease they become confluent, and at length form large, irregular patches. At first the surface of the patch is usually smooth, marked only by fine intersecting fissures, and is but slightly elevated above the surrounding mucous membrane. At this time the papillæ are often prominent and large, but, as the epithelial cells accumulate, they atrophy and are buried out of sight, and the surface may become raised several millimetres above the healthy mucous membrane. In some instances we find that a portion of the horny mass has been thrown off, and has left a central depression which may be ulcerated. These latter changes take place only in those cases which have been exposed to great irritation or which are approaching the stage of epithelioma. About the younger and growing patches we find an erythematous border, but this finally disappears. In cases associated with syphilis, cicatrices and nodosities or deep fissures of the tongue are usually found, and, in those which have passed into epithelioma, induration and thickening of the subjacent tissues, with deep, unhealthy ulcers, are likely to be present.

Diagnosis.—Leucoplakia is liable to be mistaken for

what Guinaud* has termed the "professional patches," found in glass-blowers, for "smokers' patches," mercurial patches, psoriasis linguæ, syphilitic patches, and epithelioma unconnected with leucoplakia. The "professional patches" occur only in old glass-blowers, particularly in bottle-makers, and are found symmetrically upon both sides of the mouth, on the lateral surface of the jaw, and around Stenson's duct. "Smokers' patches" are more irregular in seat than those of leucoplakia, and are commonly located near the commissures of the lips, but not upon the dorsum of the tongue or inner side of the cheek. Again, the epithelium covering their surfaces is thin and closely adherent, so that it can not be removed, as in the disease under discussion. Mercurial patches are not so thick as those of leucoplakia, are never quite white, and are found on all parts of the tongue, but particularly where it is pressed against the teeth. According to W. Fairlie Clarke,† psoriasis is an affection in which circumscribed patches of epithelium assume a white, opaque appearance, which, after a day or two, are thrown off, when the epithelium is speedily restored; but soon other patches appear and go through the same course, until, after a time, the whole surface of the tongue becomes denuded and of a uniform red color, with crescentic markings or depressions, which, it will be observed, is very unlike the course of leucoplakia. Syphilitic patches are not so white as those of leucoplakia; they are usually round or oval, and more regular in form, and they seldom occur on the cheek, but are found principally on the tip or margins of the tongue, and often on its lower surface, which is never invaded by leucoplakia. Syphilitic patches do not become so thick as those of leucoplakia, and in syphilis the lymphatic glands are soon involved, which is not the case in the latter unless it has become can-

* *Loc. cit.* † *Loc. cit.*

cerous. The pain is more severe in leucoplakia than in the syphilitic disease, and anti-syphilitic treatment causes no improvement, but may aggravate the affection. When syphilis and leucoplakia co-exist, the diagnosis is difficult, and sometimes can only be cleared up by specific treatment. Cancer arising without previous leucoplakia is distinguished from the latter by its history. In cancer, the induration of tissues and the final ulceration are not preceded by the chronic white patch, and are attended by more constant pain, with profuse salivation and a very offensive odor.

Prognosis.—The duration of the disease is uncertain. One authority mentions two cases in which cancer supervened in less than six months. Sir James Paget * mentions one case which terminated in cancer at the end of eighteen months, several of Mauriac's † were of eleven to thirteen years' duration, while of the two cases seen by Hulke, ‡ which terminated in this way, one had existed twenty years. Others have mentioned cases of thirty to forty-five years' duration. Debove # and Bazin, ‖ who make no clear distinction between leucoplakia and syphilis, and Kaposi, △ who speaks of leucoplakia as a universal product of syphilis, very naturally believe that the affection is frequent, that it does not terminate in cancer so often as we should suppose from the writings of others, and that it is very amenable to treatment. Schwimmer ◊ says that the disease is frequently transformed into malignant formations and ends fatally, and the general belief is that it is very likely to end in epithelioma. Vidal ↕ believes that one half the cases terminate in this way. On this point Mauriac ↕ says that "transformation into epithelioma has often been noted, therefore it

* *Loc. cit.* † *Loc. cit.* ‡ *Loc. cit.* # *Loc. cit.* ‖ *Loc. cit.*
△ "Syphilis der Haut," Wien, 1875, vol. iii.
◊ *Loc. cit.* ↕ *Loc. cit.* ↕ *Loc. cit.*

is important to cure the superficial affection, or it may pass into a deep, destructive, mortal disease."

The same author thinks that there are three varieties of leucoplakia (or, as he terms it, psoriasis linguæ). One, which he says is curable (due to syphilis), we do not think belongs to this class of diseases. The others he considers incurable. The variety which he attributes to the arthritic or dartrous diathesis he terms innocuous, while the third variety—the epitheliomatous—is incurable and malignant.

Among the indications that leucoplakia is passing into epithelioma are: non-inflammatory enlargement of the lymphatic glands, with exfoliation of the thicker portion of the patch, the formation of an ulcer, the supervention of sharp pain, salivation, and at length induration of the subjacent tissues. Finally, great swelling in the region of the jaw is likely to occur, and death takes place from exhaustion.

Treatment.—In cases of leucoplakia all sources of irritation, particularly those resulting from the use of tobacco and alcoholic stimulants, should be at once removed, and if the digestive organs are deranged, as is frequently the case, they should receive proper attention. Aside from these measures, most authorities believe treatment of little or no avail. Schwimmer* says that local treatment is not rational; that alkaline washes and the waters of Vichy and St. Christan, and other alkaline waters, though highly recommended by Bazin† and other French writers, are not satisfactory in their effects; and that, even if improvement does occur under their use, it is not likely to be enduring. Debove‡ and Bazin state that many cases of buccal psoriasis may be cured, but their favorable results may be due to the fact that they included many syphilitic cases in this class.

* *Loc. cit.* † *Loc. cit.* ‡ *Loc. cit.*

By an examination of Debove's recorded cases, I find that, exclusive of the syphilitics, none of the patients seem to have been entirely cured, though many were treated by both Bazin and Debove, and a number are reported as "something better" after several months of treatment. Mauriac,* who believes that there are three varieties of leucoplakia—viz.: 1, the dartrous and arthritic; 2, the syphilitic; 3, the epitheliomatous—recommends arsenic and alkalies for the first, mercury and iodides for the second, and surgical measures for the third; however, he urges great prudence in using any of these measures, and states that the internal remedies have been found almost useless, and that mercury and the iodides are dangerous in the absence of syphilis.

For local application, the caustics which have commonly been employed are nitrate of silver, chloride of zinc, tincture of iodine, and acid nitrate of mercury; but none of them seem to do any good excepting in syphilitic cases, which I do not include under the term leucoplakia. Soothing applications seem to have been most beneficial, but they give only temporary relief.

E. Vidal † says that "mercury and iodide of potassium aggravate the disease the more advanced it is," and that in the papillomatous state they precipitate epithelioma. When the affection has passed into epithelioma nothing can be of much benefit excepting thorough excision, but even this has not often been followed by happy results.

Henry Morris ‡ advises that, after other measures have been fairly tried, if the epithelium is constantly thrown off, leaving abrasions or ulcerations which are painful, the tongue should be excised without delay. He states that "the prospect of curing, or even much improving, ichthyosis by treatment is most unfavorable."

S. James A. Salter # reported a case, which seemed to

* *Loc. cit.* † *Loc. cit.* ‡ *Loc. cit.* # *Loc. cit.*

have been leucoplakia, which was cured by extirpation and cauterization of the wound with the actual cautery. In the case which I report it will be observed that internal remedies did no good, and that local applications of tincture of iodine, nitrate of silver, and the acid nitrate of mercury greatly increased the patient's sufferings, and would doubtless have aggravated the disease had they been persisted in; but, as soon as the actual (galvanic) cautery was employed, relief from all pain was obtained, and by a persistent, careful use of it the disease was eradicated.

In considering this favorable result, of course we must not overlook the reported tendency of the disease to recur; but the fact remains that the belief in this tendency is based on a study of cases in the majority of which the diseased patch was never entirely removed. In my case the mucous membrane is perfectly healthy after a lapse of four months; therefore I believe the treatment adopted for it will prove curative in many cases if applied before epithelioma has developed.

From a study of the literature of this subject, and from my own small experience, I arrive at the following conclusions:

1. Leucoplakia buccalis is an idiopathic disease, distinct from psoriasis, "smokers' patches," and syphilis. It is largely confined to men past middle life, but it occasionally occurs in women.

2. The disease is so commonly found in inveterate smokers that the abuse of tobacco may fairly be considered as an exciting cause, though cases occur where tobacco has never been used.

3. The affection is chronic and, finally, in a majority of cases, terminates in epithelioma.

4. Internal treatment and the local application of sedative, stimulant, or caustic drugs are, in nearly all cases, either

useless or injurious, and the latter are sometimes disastrous by hastening the development of epithelioma.

5. The actual cautery or the galvano-cautery will probably enable us to cure many cases if they are treated sufficiently early, provided it is applied to only a small spot at each sitting, and carefully, so as not to destroy the healthy tissues beneath the changed epithelium.

A Typical Case.—L. C., aged forty-three, printer, came to me in September, 1884, complaining of a sharp pain in the left side of the mouth, particularly when eating. This, he said, began two years before, when he had a tooth drawn. Two months later he first noticed a small white patch on the buccal mucous membrane. Upon examining the mouth, I found a large, irregular, milk-white patch, extending from the border of the gums down through the gingivo-buccal groove, upward along the left cheek, and from the first bicuspid tooth, in front, to the last molar, behind. Altogether this was as large as a silver dollar; its surface was marked by fine intersecting lines, which divided it into numerous polygonal spaces.

At the center of the patch, on the buccal surface, was a depression about 3 mm. in depth and 1 cm. in diameter, surrounded by elevated, hard margins, which gradually became thinner toward the edge of the patch. This margin was deeply fissured in two or three places. About half an inch in front of the anterior portion of this patch, on the under lip, was a small, white, warty growth, about 4 mm. in height by 3 mm. in diameter.

A critical investigation of the case failed to discover any history or signs of syphilis or hereditary disease. The patient chewed tobacco to excess and had smoked occasionally. He stated that previous to the removal of the tooth, and until the pain became troublesome, he had been accustomed to carry his quid of tobacco in the left side of the mouth, at the location of the patch. His general health was perfect, and he had never suffered from rheumatism or eruptive diseases, and did not use alcoholic stimulants. During the first few months of the affec-

tion pain came on about once in two weeks and would last several hours, but the attacks gradually became more frequent, until seven or eight weeks before I saw him, during which time the pain had been constant and often severe. The patch had been cauterized occasionally with nitrate of silver, but was never benefited.

When I first saw the case, suspecting that it might be syphilitic, I ordered iodide of potassium in large doses and directed that tobacco be discontinued. I cauterized the patch with tincture of iodine, but the application caused such severe and protracted smarting that I began at once to be doubtful about the character of the disease. Three days later I applied the solid nitrate of silver to a small portion of the patch, with similar results, and four days later the acid nitrate of mercury. This last application caused intense pain that lasted about seven hours, which, he said, drove him nearly crazy. I was now convinced that the disease was not syphilitic, and had the patient see Professor Hyde, who pronounced it leucoplakia. I next cauterized the central depressed portion of the patch with the galvano-cautery, which caused momentary smarting, but perfectly relieved the severe pains from which he had suffered for weeks, and they never returned. Two days later I destroyed the small, wart-like projection in front of the large patch, and subsequently, about every fifth or sixth day, I cauterized a small spot, nearly a centimetre in diameter, at the edge of the patch. These cauterizations were so superficial that only the epithelium was destroyed, the membrane beneath being but slightly burned; indeed, so light were they that a few times I was obliged to again cauterize the same spot before all the epithelium was destroyed. I found that as these cauterized places healed the mucous membrane appeared natural, and finally, after about thirty applications of the cautery, the entire surface presented the appearance of healthy mucous membrane, showing only two or three small cicatrices, where the cauterizations had been unusually deep. The iodide of potassium, which did no good, was discontinued soon after I began the use of the galvano-cautery. Now, at the end of four months after the last cauterization, the mucous membrane remains perfectly healthy.

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